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Male fertility in cystic fibrosis.

Sanjay H. Chotirmall
Beaumont Hospital, Dublin

Amandeep K. Mann
Royal College of Surgeons in Ireland

Peter Branagan
Beaumont Hospital, Dublin

Cassandra O'Donohoe
Beaumont Hospital, Dublin

Anne M. Lyons
Beaumont Hospital, Dublin

See next page for additional authors

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Authors
Sanjay H. Chotirmall, Amandeep K. Mann, Peter Branagan, Cassandra O'Donohoe, Anne M. Lyons, Maura G. Flynn, Cedric Gunaratnam, Shane J. O'Neill, and Noel G. McElvaney

G McKenna, PF Allen, D O'Mahony, C DaMata, M Cronin, N Woods
Department of Restorative Dentistry, Cork University Dental Hospital, Wilton, Cork
Tel: +353 21 490 1100 ex 5033
Fax: +353 21 454 1193
Email: g.mckenna@ucc.ie

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Male Fertility in Cystic Fibrosis

SH Chotirmall, AK Mann, P Branagan, C O'Donohoe, AM Lyons, MG Flynn, C Gunaratnam, SJ O'Neill, NG McElvaney
Respiratory Research Division, Education & Research Centre, Beaumont Hospital, Beaumont Road, Dublin 9

Abstract
Infertility rates among males with cystic fibrosis (CF) approximate 97%. No information is currently available within Ireland determining an understanding of fertility issues and the best methods of information provision to this specialized group. This study aimed to determine understanding and preferred approaches to information provision on fertility issues to Irish CF males. A Descriptive Study utilizing prospective coded questionnaires was mailed to a male CF cohort (n=50). Sections included demographics, fertility knowledge & investigation. Response rate was 16/50 (32%). All were aware that CF affected their fertility. More than two-thirds (n=11) were able to provide explanations whilst only one-third (n=5) provided the correct explanation. Significant numbers stated thoughts of marriage and a future family. Half have discussed fertility with a healthcare professional (HCP). Mean age of discussion was 21.9 years. One third preferred an earlier discussion. The commonest first source for information was written material which was also the preferred source. Three-quarters requested further information preferring again, written material. Significant gaps in sex education of Irish CF males exist. Discussion should be initiated by HCPs and centre-directed written material devised to address deficiencies.

Introduction

The Irish population has both the highest incidence[1] (2.98/10,000 individuals) and carrier rate (5.26%) of CF worldwide[2]. With improving disease survival[3,4] patients are increasingly considering "getting married" and "having children". This has significant implications for CF services. It remains the CF team's responsibility to provide adequate and accurate up to date fertility information to our CF patients even before we are asked 'the difficult questions'. At present, there is no available information in the Republic of Ireland illustrating the knowledge of fertility issues among Irish males with CF. Without such information, it is difficult to address and subsequently plan the optimal methods to deliver such information to this specialized group. Infertility in males with CF was first established in the 1960s[5-7] and rates approximate 97% attributed to azoospermia. Despite this, new techniques are enabling males with CF to father children. Small but significant numbers of male CF patients remain fertile. This is observed in particular genotypes[8,9] serving to re-emphasize the importance of fertility information provision in CF.

Most studies have assessed infertility and parental rates in CF. To a lesser extent has research been conducted to assess understanding of fertility issues and the preferred modes of communication for information delivery. Hames[10] and Nolan et al[11] illustrated that males with CF were grossly unaware about the reproductive and sexual complications associated with the disease. Subsequent work[12-14] has shown that although awareness has improved, they still remain ill-informed in terms of the broader aspects of reproductive education. Consistently, there have been calls for improved information provision[15]. Noteworthy questions remain. Are male CF patients aware of their infertile status? Who tells them? When are they told? When should they be told? When do they want to be told? In this study, we determine the current understanding and knowledge of fertility issues among Irish CF males. We also determine the desired approach to fertility information and investigation provision during routine CF care.

Methods

A prospective coded descriptive study was conducted (2006-07)

utilizing a ten-page self-rated questionnaire. Sections included demographics, CF status, lifestyle, personal relationships and fertility knowledge and investigations. A systematic random sample of a male CF cohort attending our centre were included (n=50). Approval for the study was obtained from our institutional ethics committee and all analyses performed using SPSS Version 16.0.

Results

Fifty CF males were included with a response rate of 32% (n=16) commensurate with similar studies. Mean age was 24 years old (range 19-35, SD+/- 3.98).

Lifestyle Issues

50% of responders were in full-time (n=6) or part-time (n=2) employment. One-quarter (n=4) were unemployed and a further quarter 'students'. Almost all responders (87.5%, n=14) were non-smokers but consumed alcohol. Mean weekly units consumed was 11.1 (SD+/-15, range 1-60). Almost half (43.8%, n=7) admitted illicit drug use and of this group, three admitted regular use.

CF Status

Mean age at diagnosis of CF was 2.47 years (SD+/- 5.47, range 0-22). Over one-third (37.5%, n=6) had a sibling with CF. Half (n=8) had FEV1 >70% predicted. Three patients have been referred for lung transplantation. Most (81.3%, n=13) were aware of their sputum growths. Mean number of infective exacerbations over the preceding year was 3.34 (SD+/-3.06) and mean hospital admissions 1.56 (SD+/-2.19). Six responders (37.5%) had CF related diabetes (CFRD). Self-reported compliance was good with 81.3% (n=13) reporting 'all times' or 'most times' compliant with prescribed therapies. Half (n=8) were aware about the availability of genetic counselling.

Knowledge of Fertility Issues

All responders were aware that CF affected their fertility however 68.8% (n=11) were aware of an explanation of which only 31.3% (n=5) were correct. Almost all stated knowledge of a definition for the term 'infertility' (93.8%, n=15) however only 56.3% (n=9) were able to explain it. Conversely for the term 'impotence' 68.8% (n=11) claimed knowledge and all were able to explain it. When asked to estimate expected percentage of infertility among male CF patients, a mean value of 89.0% (SD+/-19.9, range 20-99) was obtained.

Personal Information & Relationships

Nearly all studied (87.5%, n=14) were single. However, of this group, six were in a relationship. Two patients (12.5%) were married. A proportion stated that CF interfered with relationships (43.8%, n=7) and the most cited reason was the shortened life expectancy (31.3%, n=5). The next most cited reason was frequent hospital admissions. The majority are sexually active (87.5%, n=14) and three-quarters (n=12) have more than one partner. Mean life-time partner number was 4.08 (SD+/-3.87, range 2-16) and mean age of first sexual intercourse was 17.8 years (SD+/-2.02, range 15-22). Barrier protection was only used by half (n=8). In this group, most (62.5%, n=5) cited both contraception and the prevention of sexually transmitted diseases (STD) for using condoms. Significant proportions of the cohort had thoughts of 'marriage' (68.8%, n=11) and 'starting a family' (56.3%, n=9). One-quarter had been in a relationship with another individual with CF and none of those studied had children.

Only half (n=8) had ever discussed fertility with a HCP and of this group, half were self-initiated. Only 25% of all discussions were initiated by the HCP. Another group of initiators were parents (12.5%). Mean discussion age was 21.9 years (SD+/-2.6, range 18-26). More than one-third of those who had discussions (37.5%) preferred an earlier discussion. Most (87.5%) individuals were satisfied with the information provided during discussions and few (12.5%) left with 'unanswered questions'. The terms 'infertility' and 'impotence' were discussed in 56.3% (n=9) and 25.0% (n=4) of cases respectively. The mean age preference to be told of fertility status was 16.9 years (SD+/-2.24, range 12-21). The commonest first source of hearing of infertility was written material (43.8%, n=7) which in 68.8% (n=11) was the preferred source. Seven patients (43.8%) admitted having further fertility questions but being too embarrassed to ask them. Knowledge of an infertile status only affected relationships in a small number of cases (12.5%).

Fertility Investigations

Three-quarters (n=12) requested more information with the preferred mode being written material (68.8%, n=11). Two patients had semen analysis and almost everyone (93.8%, n=15) would accept an opportunity for this if offered. When asked the ideal time to perform such analysis, a mean age of 18.9 years (SD+/-2.45, range 16-25) was suggested.

Discussion

This study, the first in Ireland describes data pertaining to knowledge and understanding of male infertility in CF. Considering the sensitive and personal nature of questions asked, the moderate response rate was comparable with prior studies. The results presented represent useful information but must be interpreted with caution in terms of the limited response rate.

All studied were aware that CF affected their fertility however most did not know the frequency or reasons for this. This awareness is an improvement on earlier studies[10,16] showing that 27% of CF males knew of their infertile status. Such findings are contrasted by Sawyer[12] and Fair et al[14] which both established >90% awareness. Such discrepancy has been attributed to the varied ages of patients studied rather than an improvement in education. Awareness of infertility takes on greater importance as one gets older and consequentially awareness parallels age. An important point to consider, highlighted by our study, is the distinction between "awareness" and "understanding". In our study, although all patients were 'aware' of infertility, most did not 'understand' it. This is demonstrated by the inability to provide correct explanations of infertility although admittingly being aware of it. Similarly, most claimed an understanding of terminology such as 'infertility' or 'impotence' but again could not provide definitions when questioned. Hull et al[17] have previously shown that 'reproductive counselling' is not a priority within the worldwide CF healthcare structure. This is reflected by the disparity identified between 'awareness' and 'understanding'. CF services should take responsibility for making patients 'aware' but enabling them to 'understand' fertility concepts.

Many were having intimate relationships, and thoughts of marriage, and having families were considered by half. Shepherd et al[18] described that CF patients were as likely as the normal population to be married or in a relationship, a statement concurrent with our findings. In view of this, it is imperative that CF patients are educated appropriately about the impact of the disease on relationships, sexual life and health. None evaluated had children and although almost all reach reproductive age, it has been found that few actually have children and even fewer seek consultation on assisted fertility techniques[19]. Consequently, although definite thoughts about having children exist, it remains unclear to what extent this translates into actually becoming a parent. It may well be the lack of information provision, education, advice and direction during CF care that precludes patients from believing that despite their infertile status, there are techniques available enabling them to successfully father children. Significant numbers are sexually active with intermittent usage of contraception. Our study has additionally uncovered multiple partners among sexually active CF males. This consequentially has downstream effects on sexual health and implications for CF services.

The numbers discussing fertility with a HCP are suboptimal and

half were self-initiated. Such findings were reproduced by Fair et al[14] who showed that most never discussed fertility with a HCP and, of those that did, the majority did not find out what they wanted. This last point differed in our findings. Within our cohort most were satisfied with both the discussion and information provided but felt it was happening much later than preferred. Reported barriers to discussion[20] include lack of training, embarrassment and difficulty finding the "right-time" for discussion. Recent work by Frayman et al[21] has additionally identified that whilst parents are well informed of fertility issues in CF, there is limited opportunity for discussion with a HCP.

Optimal timing, age and adjuvant modes of communication for discussion are debated. In our study, we identified a preferred mean age of seventeen concurrent with prior work[10,12,14,22,23] and the preferred source written material. Although controversy remains with regard to 'the right' age to introduce such concepts, what remains undoubted is the importance of making any educational intervention culturally and contextually-appropriate for patients. Additionally, it is imperative that discussion concerning fertility begin prior to transfer to an adult care unit. An initial discussion during the latter stages of paediatric care and again subsequently following transfer to adult care should take place. This second discussion aims to reinforce factual concepts introduced during paediatric care but additionally to outline the available fertility investigations and techniques that patients can avail of.

Few have had fertility investigations (semen analysis) performed but significant interest in this was discovered through our study. Most requested this to be offered routinely as part of CF care concurrent with data from Rodgers[22] and Sawyer et al[15]. Overall, our study has identified both a need and desire for more fertility information provision and discussion among CF males. Written material was highlighted as the most desired mode of communication and subsequently developed at our centre to tailor deficiencies identified through this study. This material is available through the Cystic Fibrosis Association of Ireland (CFAI). The booklet entitled 'Sexual Health in Cystic Fibrosis' covers short topics addressing both male and female sex issues. Such material differs from others in that its development has been driven by deficiencies identified by this study. It remains important to highlight that whilst written material was our samples 'preferred' method, it remains questioned whether this is actually the 'best' method for education. A combination of written material and discussion with a HCP may provide most benefit.

We have identified significant gaps in fertility education among a cohort of Irish CF males. Discussions should be initiated to aid understanding at the earliest opportunity. We recommend two separate formal discussions about fertility supplemented by written material – one in paediatric care and then a second after transfer to an adult unit. The initial discussion should contain mainly factual information. The next discussion should take place soon after transfer of care to an adult CF facility. It should reinforce earlier factual content but additionally contain information on fertility investigation, provision of semen analysis if opted for and subsequent referral if desired to a specialist fertility clinic. Appropriate and directed written material similar to that devised at our centre can be used adjunctively to address identified deficiencies.

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Correspondence: SH Chotirmall
Respiratory Research Division, Education & Research Centre, Beaumont Hospital, Beaumont Road, Dublin 9
Email: schotirmall@rcsi.ie

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